Title: Patient and health care impact of a pilot rheumatic heart disease screening program

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ABSTRACT AND KEYWORDS

Aim: To assess the impact of a pilot screening program for rheumatic heart disease (RHD) on patient quality of life (QOL) and health services.

Methods: A proxy-respondent QOL questionnaire (CHQ-PF28) was used in children with a potentially abnormal screening echocardiogram and in matched normal controls. The health services response and impact on health services was evaluated using medical record review, responses from carers and an online survey of health care providers.

Results: QOL was assessed in 68 children. Potentially abnormal screening echocardiograms were associated with poorer QOL in the General Health Perception \((p<0.05)\) and Parental Impact – Emotional \((p<0.05)\) domains. Health services notified 82\% of children and clinical review occurred in 56\%. Only 64\% of contacted carers were able to recall that a potential abnormality was detected. A potentially abnormal echocardiogram was associated with a change in management in 6\% (2/34) of children. When surveyed, 49\% of health providers were aware of the RHD screening program, 29\% had seen children referred with screening abnormalities and 85\% of these providers stated this had an impact on local health care delivery.

Conclusion: This pilot RHD screening program was associated with poorer child and carer QOL, greater health provider workload and suboptimal clinical follow-up with little change in clinical management. The adoption of ongoing screening for RHD in high-risk populations should be approached cautiously. Further research is required to facilitate the development of improved echocardiographic diagnostic criteria for RHD and the systematic assessment of the benefits and adverse effects of such screening.

Key words: Screening; rheumatic heart diseases; psychosocial impact; health services evaluation; quality of life; echocardiography.
KEY POINTS – WHAT IS ALREADY KNOWN ON THIS TOPIC

1. Rheumatic heart disease (RHD) remains a health issue for Aboriginal Australian and Torres Strait Islander peoples
2. Detection of the earliest changes of RHD provides an opportunity to implement secondary antibiotic prophylaxis
3. A significant proportion of echocardiography confirmed RHD is not associated with a murmur on auscultation or a history of ARF

KEY POINTS – WHAT THIS PAPER ADDS

1. Screening for RHD in high risk populations is feasible
2. Screening for RHD can have an adverse impact on a child’s quality of life and can place a significant impact on local health services
3. Improved diagnostic criteria to differentiate the earliest echocardiographic changes of RHD from normal findings are required
Introduction

Acute rheumatic fever (ARF) is a non-suppurative sequelae to group A beta-haemolytic streptococcal infection. Repeated episodes of ARF can induce a cycle of carditis and valve scarring and dysfunction resulting in rheumatic heart disease (RHD). Early detection of mild RHD presents an opportunity for intervention with secondary antibiotic prophylaxis that can in turn limit further valve damage.

The presence of RHD is not always associated with a history of ARF or a murmur on auscultation. Detection of those at risk of developing significant RHD is therefore difficult based on history and clinical examination alone. With the development of portable and relatively affordable echocardiography there has been increasing interest in using echocardiography screening to identify early RHD in children from populations at high risk of ARF/RHD. Indeed the Australian guidelines for the diagnosis and management of ARF/RHD recommend screening programs to detect undiagnosed RHD in high risk populations, wherever this is possible. Nonetheless the utility of current echocardiographic diagnostic criteria for RHD and normal findings in this setting and the impact of echocardiographic screening on children, families and health care services remain poorly understood.

A northern and central Australian RHD echocardiographic screening and prevalence study of over 5000 predominantly Aboriginal and/or Torres Strait Islander children aged 5-14 years was implemented between 2008 and 2010. To facilitate clinical follow-up of potentially abnormal studies early reporting of these screening echocardiograms was undertaken by cardiologists with experience in RHD and clinical practice in this setting.
The aim of this study was to determine the impact of a potentially abnormal clinician reported screening echocardiogram on children, families and health care systems and whether this in turn translated to clinical follow-up and a change in management.

Materials and methods

The study involved the assessment of three aspects of this pilot screening program: child quality of life (QOL); health service response; and health service impact. This study occurred at least 10 months following the completion of the earlier screening project.

The assessment of QOL was undertaken in Aboriginal Australian and/or Torres Strait Islander and non-Indigenous Australian children found to have a potential abnormality on a clinician reported echocardiogram and in age, gender, ethnicity and location matched children with a screening echocardiogram reported as normal. Children resided in one of three north Queensland sites: Yarrabah; Cairns and Thursday Island (Figure 1).

Demographic data relating to children and parents and QOL was collected using the principal carer as the informant. A 28-item study specific questionnaire assessed awareness of the echocardiogram result, medical follow-up and sociodemographic factors including location, ethnicity, income, carer level of education and child comorbidities. Comorbidities were defined as any condition(s) requiring ongoing medication use or regular clinical follow-up. The CHQ-PF28 (HealthActCHQ Inc., Cambridge, MA) was used to assess QOL. It is a validated parent reported questionnaire for children aged 5-18 years. Whilst it has been validated in an Australian setting, this was its first documented use in Aboriginal Australian and Torres Strait Islander
populations. The CHQ-PF28 assesses 14 physical and psychosocial concepts with two associated summary scores. Concepts explored include the well-being of the child and the impact of the child's health on parents' QOL and the family. Each domain score is expressed as a number from 0 to 100 with 0 indicating poor and 100 excellent health and functioning. All carers were contacted by telephone and the questionnaire was completed verbally with one of the researchers (EKW).

The assessment of the health provider perceived impact of echocardiographic screening on their local health service was undertaken using an internet based survey (Survey Monkey Online Survey Software, Palo Alto, CA). Primary or specialist health care providers supplying care to Queensland sites where the earlier screening study was undertaken; or located in a north Queensland cardiology or paediatric tertiary referral centres were invited by email to participate. A reminder email was sent two and six weeks following initial contact. Returned surveys were excluded from analysis if fewer than 50% of questions were completed or if they were completed by staff who were not directly involved in clinical care.

Clinical follow-up, eventual diagnosis and alteration in management was determined by combining information from the parent questionnaire (see above) and review of the child’s local primary health care and regional paediatric outreach service health record.

Data analysis was undertaken using IBM SPSS Statistics (version 19.0, SPSS Inc., Chicago, Il). All tests were two-sided, and a p-value of <0.05 was considered to be statistically significant. Group comparisons of categorical data were made using Pearson’s $\chi^2$ or Fisher’s exact test. CHQ scales were reported as continuous variables, and multivariate analysis of variance was performed to adjust for confounding factors.
Qualitative data derived from the health provider survey was thematically organised according to key words or statements. Pattern recognition was used to organise the themes into negative and positive impacts on the health service.

The study was approved by the Cairns and Hinterland Health Service District and the James Cook University Human Research Ethics Committees. Written informed consent to complete the questionnaires and health record review was obtained from all carers. Consent from health providers for the on-line questionnaire was inferred by completion.

Results

QOL and a potentially abnormal echocardiogram

Sixty-eight primary carers of children who had previously been enrolled in the earlier screening and prevalence project participated in this study. Demographic characteristics are summarized in Table 1. Carers of children with a normal echocardiogram reported a higher level of education compared with carers of children with a potentially abnormal echocardiogram.

Of the children with a potentially abnormal echocardiogram, 32 (94%) demonstrated subjective valvular thickening, 32 (94%) valvular regurgitation, 8 (24%) were assessed as having possible or probable RHD and 3 (9%) were diagnosed with congenital heart disease.

QOL subscales for children with a potentially abnormal and normal echocardiogram are presented in Figure 2. Given carer level of education was associated with poorer QOL this was controlled for in multivariate modeling. After controlling for this there was a persisting independent association between the presence of a potentially abnormal
echocardiogram in a child and poorer QOL in the subscales General Health Perception ($F(1)=4.283$, $p<0.05$, partial $n^2=0.062$) and Parental Impact – Emotional ($F(1)=4.675$, $p<0.05$, partial $n^2=0.067$).

**Health service response to abnormal screening results**

Local health care providers recorded contact with 28/34 (82%) of carers of children who had a potentially abnormal screening echocardiogram. Face-to-face clinical review with a primary health care provider or paediatrician had occurred in 19/34 (56%). Of the 28 carers who had documented contact with local health care providers only 18/28 (64%) could recall that their child’s echocardiogram result may have been abnormal. Although 34 children involved in this study were thought to have a possible abnormality only one of these children was determined to have definite RHD on medical review and commenced on penicillin prophylaxis. Additionally, another child with incidentally detected congenital heart disease was referred to a tertiary cardiology service.

**Health provider perspective of echocardiogram screening for RHD**

Of the 194 health providers who completed the online questionnaire 115 respondents met inclusion criteria. Respondents who met inclusion criteria included indigenous health workers (24, 21%), nurses (57, 50%), general practitioners/district medical officers (10, 9%), paediatricians (12, 10%), cardiologists (8, 7%) and echocardiographers (4, 3%). Of these, 56 (49%) were aware of the earlier screening project and 33 (29%) had seen children referred with potential abnormalities.

Of those who had seen children referred with screening abnormalities, five (15%) stated that there was no impact on their health service, 13 (39%) saw referrals with minimal impact, five (15%) with moderate impact, eight (24%) with high impact and two (6%)
Health providers stated that they were completely inundated and unable to keep up with referrals.

Health provider comments regarding the health service impact particularly dealt with the limited capacity to deal with a significant number of paediatric cardiology referrals. The difficulty in interpreting borderline or minor echocardiography changes was also highlighted with one provider noting that it was

“...near impossible to make diagnostic decisions based on an echocardiogram result not conducted by yourself on a child of which you do not know the clinical background...”

The inability of local health record systems to facilitate easy access to a child’s past medical history and investigations to ascertain the likelihood of a possible prior episode of ARF was an additional source of frustration for local providers. Whilst respondents noted the program improved health awareness, facilitated engagement between the community and local health service, and raised the profile of ARF/RHD in the community a recurring theme was the uncertainty engendered by an echocardiogram result than may, or may not, be normal. As noted by one local health care provider

“Telling patients that their child had an abnormal result, and then trying to explain that it might be within the normal range was very stressful...”

DISCUSSION

This study is the first to assess the impact of echocardiography based screening for RHD on children and carers and the response and impact on local health care services. It has also demonstrated that an established QOL assessment tool has utility in an
Aboriginal and Torres Strait Islander setting and that carer education, in addition to screening, can influence this. Whilst ARF and RHD remain important contributors to Aboriginal and Torres Strait Islander health disadvantage the findings of this study would indicate that at present the implementation of echocardiographic screening for RHD, outside the confines of research, should be deferred. Whilst such screening may have a future role, diagnostic criteria need to be validated and standardized and the benefits of screening must be demonstrated to be superior to the potentially adverse effects on children and health services.

Earlier research-based screening for RHD has demonstrated that many children with changes on echocardiography consistent with RHD do not have a audible murmur on auscultation and up to 43% of patients with RHD do not have a history of ARF. Given the demonstrated efficacy of secondary antibiotic prophylaxis in preventing ARF, and presumably the progression of RHD, a persuasive argument may be mounted to support echocardiography based screening for RHD in high risk populations. Nonetheless the adverse effects of screening programs are not commonly acknowledged and a number of these are highlighted by this study.

Our findings highlight similar issues seen in earlier reports of health related screening in children. Evaluation of the impact of screening for hypercholesterolaemia in otherwise well children demonstrated anxiety and over-protectiveness in parents of those children found to have an abnormal result. Such adverse affects of screening can also persist after secondary testing has demonstrated no potential abnormality. In parents of children with a false-positive neonatal screening result the adverse effects on mood and behaviour persisted despite subsequent reassurance and counseling.
More specifically the impact of earlier auscultatory screening for cardiac disease in children has demonstrated similar adverse effects on children and parents. In Bergman et al’s study they identified a link between false positive screening results and parental anxiety, concluding that parents worry “...in children suspected of having cardiac disease, that catastrophe is apt to be perceived as a sudden heart attack”.

Despite age, gender, ethnicity and location matching, carers of children with a potentially abnormal echocardiogram demonstrated significantly lower levels of formal educational attainment. This was in turn associated with lower carer reported QOL. Whether lower carer education, an increased risk of a potentially abnormal screening echocardiogram and poorer QOL are causally linked or confounded remains to be seen. Nonetheless the persisting difference in QOL between groups when controlling for carer education would support a independent relationship between the presence of a potentially abnormal screening echocardiogram and poorer QOL.

The use of a QOL tool in an Aboriginal and Torres Strait Islander setting has not previously been reported. Whilst this may raise issues of validity and reliability our findings, where we have demonstrated the ability to detect plausible differences in QOL, would support its utility in the future evaluation of health care interventions in Aboriginal Australian and Torres Strait Islander peoples.

This study sought to not only evaluate the impact of echocardiographic screening on carers and children but also to determine how this translated to health care delivery and the perceived impact on local health care services. Whilst potentially abnormal screening echocardiograms had generated an initial health service response this was associated with a lower level of face-to-face clinical review. The disparity between
identification at the local health service level and subsequent clinical review may in part be explained by the lack of carer recall of the echocardiogram findings despite documented contact by the local primary health care service. It was beyond the scope of this project to map and critique the systems underlying primary and specialist health care information handling, patient registration and recall. Nonetheless the relative disconnect between registration (as demonstrated by the initial clinical response), recall (clinical review) and client understanding (carer knowledge of diagnosis) serves to highlight the importance of developing and supporting effective health care systems as part of screening program implementation and evaluation.

Any health related screening will have an impact on health care delivery. This can occur at multiple levels including patient counselling regarding results and management, providing and interpreting secondary investigations to confirm or refute a screening-based diagnosis and management of those confirmed to have the condition of interest. Whilst the earlier RHD screening study occurred in consultation with primary and specialist health care providers there was not provision on the part of the study or the local health service to allocate additional resources to facilitate clinical follow-up. It is therefore not surprising that the majority of health care providers surveyed identified at least some impact on their local health care service. The fact that 30% of those surveyed who were involved in this follow-up felt this impact was substantial (high or completely inundated) emphasises the importance of clearly assessing and resourcing the increased health service activity associated with any planned screening program prior to implementation. The significant impact on local service activity does not even then capture the full health service impact of screening. The feedback provided by health care providers highlighted that, in addition to being busier, such screening and
particularly the uncertainty associated with interpretation was stressful. In a remote
Australian setting where primary health care workforce stress and staff retention
remains a perennial issue such additional strain is obviously best minimised.\textsuperscript{13}

In conclusion based on the findings outlined a good case cannot, at this time, be made
for RHD echocardiographic screening in this Australian health care setting. These
findings do not preclude the future use of effective RHD screening. Rather such
deployment needs to be contingent on improved systems for diagnosis and clinical
follow-up, adequate resourcing of the anticipated additional load placed on local health
services and a clear demonstration that the benefits of early intervention outweigh the
potentially adverse effects on children, carers, health care providers and health care
services demonstrated here.

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FIGURE LEGENDS

Figure 1  Northern Queensland study sites

Figure 2  Carer reported quality of life (CHQ-PF28) domain and summary scores (SEM) in children with a potentially abnormal echocardiogram compared to those with a normal echocardiogram (* p<0.05 controlling for carer education)
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Table 1  Sociodemographic characteristics of study population

(n - number; IQR - interquartile range; * p<0.05)
FIGURES

Figure 1
Figure 2

CHQ-PF28 Domain and Summary Score (mean, SEM)

- Abnormal echocardiogram
- Normal echocardiogram